

Figure 2. Portal tract showing a mixed inflammatory infiltrate (lymphocytes, eosinophils, neutrophils), oedema, bile duct proliferation and damage. H&E, $\times 144$

Discussion

Captopril-induced cholestatic liver injury has been well documented⁵, but hepatotoxicity with enalapril is extremely rare. We consider that it is highly likely that enalapril was the causative agent in view of the temporal relationship of the clinical and biochemical abnormalities to initiation and cessation of treatment with the drug. The onset of jaundice was noted six weeks after initiation of enalapril therapy and its cessation was rapidly followed by a resolution of the jaundice and biochemical abnormalities.

There is one previous case of probable enalapril-provoked cholestatic liver injury³ in which a mild increase in the

transaminases and a marked rise in the alkaline phosphatase occurred without hyperbilirubinaemia. Liver biopsy revealed parenchymal damage with portal tract fibrosis. Acute hepatitis following enalapril therapy has also been reported⁴ and another case with hyperbilirubinaemia was thought to be attributable to bacteraemia and not directly to enalapril⁶.

Although cholestatic jaundice is a well documented complication of the related compound captopril, we believe this to be the first case associated with enalapril. Although many of the adverse effects associated with captopril are attributed to its sulphydryl moiety², this case of intrahepatic cholestasis related to the non-sulphydryl compound enalapril suggests that other molecular determinants are involved.

References

- 1 Jaffe IA. The adverse effect profile of sulphydryl compounds in man. *Am J Med* 1986;80:471-6
- 2 Jenkins AC, Dreslinski GR, Tadros SS, Groel GT, Fand R, Herczeg SA. Captopril in hypertension - seven years later. *Cardiovasc Pharmacol* 1985;7(suppl 1):596
- 3 Mikloweit P, Bienmuller H. Intrahepatic cholestasis induced by flecainide and enalapril. *Internist* 1987;28:193-5
- 4 Lunel F, Gripon P. Hepatite aigue apres la prise de maleate d'enalapril. *Gastroenterol Clin Biol* 1987;11:174-5
- 5 Rahmat J, Richard MD, Gelfand L, Gelfand MC. Captopril associated cholestatic jaundice. *Ann Intern Med* 1985;102:56-8
- 6 Adriansson MD. Treatment with enalapril caused leucopenia with sepsis. *Lakartidningen* 1987;84:1270

(Accepted 26 June 1989)

The modern Munchausen syndrome

M W Cooke FRCS R H Grace FRCS

Department of Surgery, New Cross Hospital,
Wednesfield Road, Wolverhampton,
West Midlands WV10 0QP

Keywords: Munchausen syndrome; HIV infection; rectal bleeding

In 1986 Robinson¹ and Miller² both described patients fabricating a history of AIDS to obtain admission to hospital. There are, however, no reports of Munchausen's syndrome³ in patients known to be HIV positive.

Case report

A 28-year-old man presented with a 24-h history of rectal bleeding of sufficient quantity to produce a pool of blood in his bed. He did not give any other history of gastrointestinal disease but said that he had lost two stone in weight.

He volunteered the information that he was sero-positive for the human immuno-competence virus. He expressed concern that he was developing acquired immune deficiency syndrome (AIDS) in his bowel as he had already had radiotherapy for a Kaposi's sarcoma on his right elbow. He had had Hodgkin's disease 5 years previously, which had required diagnostic lymph node biopsies from his neck and was followed by chemotherapy. A recurrence in his caecum required local excision. Treatment was undertaken in Paris where his parents were resident, he failed to attend for follow-up at the Royal Marsden Hospital.

Examination revealed two mature scars on his right elbow but with no evidence of radiotherapy. He had a long scar

over the anterior border of his left sternomastoid and one scar in the right iliac fossa. He had tenderness over the left side of his abdomen. Otherwise examination was normal.

Overnight he had several minor rectal bleeds, while straining to have his bowels open. Unfortunately they were never seen by any staff but he did demonstrate small amounts of blood on his clothing. Sigmoidoscopy revealed a single minor tear of the rectal mucosa and smaller tears at the anal margin. When confronted with the idea that the rectal bleeding might be self induced, he initially appeared upset but within one hour he had taken his own discharge from hospital. On Monday of the following week he presented to a local general practitioner requesting a prescription for morphine. On Wednesday he arranged an appointment in the Radiotherapy Department and on Friday presented in the Accident & Emergency Department with a history of haematemesis.

His hospital notes revealed several medical admissions within our district with attempted suicide. He had also gained admission under the care of psychiatrists with threats of suicide and a diagnosis of Munchausen's syndrome was made. Prior to these admissions, he had attended medical outpatients for extensive investigation of abdominal pain when no cause was found; it was at this time that he was found to be HIV positive.

Subsequently he was found guilty of robbery of a post office; he told police that he needed the money to buy heroin to take his own life. He created legal history when the Court of Appeal reduced his sentence from 7 to 4 years because a terminally ill person should be allowed 'a measure of hope and to improve his lot'.

Discussion

This combination of Munchausen's syndrome and HIV positivity presents hazards both for the hospital and the doctor. He has a high risk of developing AIDS however his symptoms may be attributed to his Munchausen's syndrome,

0141-0768/90/
040272-02/\$02.00/0
© 1990
The Royal
Society of
Medicine

but he cannot be investigated fully at every attendance. Any investigation in patients who are HIV positive are, by definition, kept to a minimum, particularly when they involve blood tests and endoscopy. It is therefore possible that a patient with Munchausen's can prolong his stay in hospital simply by informing the admitting doctor that he is HIV positive.

This patient was a significant hazard to staff. He was actively bleeding and took no precautions to contain the blood. On this admission he was known to be HIV positive but at all his medical consultations the following week, he failed to reveal this fact.

This case suggests the need for a register of patients who are HIV positive and have Munchausen's syndrome in order to protect hospital staff but the practicalities of absolute

confidentiality, at present, prevent this. This case also emphasizes the importance of taking simple precautions to avoid contact with blood in all patients and not only those known to be at high risk.

References

- 1 Robinson EN, Latham RH. A factitious case of acquired immuno deficiency syndrome. *Sexually Transmitted Diseases* 1987;1:54-7
- 2 Miller F, Weiden P, Sachs M, Wozniak J. Two cases of factitious acquired immuno-deficiency syndrome (letter). *Am J Psychiatry* 1986;143:1483
- 3 Asher R. Munchausen syndrome. *Lancet* 1951;i:339-41

(Accepted 10 February 1989)

Parkinsonian tremor and Waldenstrom's macroglobulinaemia

S MacRury MB ChB MRCP¹ R Watkins MD MRCP²
K R Paterson MB ChB MRCP¹ ¹Medical Unit and
²Department of Haematology, Royal Infirmary,
Glasgow G4 0SF

Keywords: macroglobulinaemia; tremor

A syndrome of hyperglobulinaemia and central nervous system affection was first described by Bing and Neel in 1936¹, but Waldenstrom's macroglobulinaemia, a condition of plasma and lymphoid cell proliferation resulting in IgM paraproteinaemia and diffuse plasmalymphoid infiltrates, was not described until 1944². Central nervous system involvement (Bing-Neel syndrome) has been found in about 25% of patients with macroglobulinaemia and usually presents as stroke, encephalopathy, myelopathy or peripheral neuropathy³. We report on a patient who presented initially with an isolated extrapyramidal lesion and was subsequently found to have macroglobulinaemia.

Case report

A 56-year-old seaman presented with sudden onset bilateral tremor of the hands at rest which had remained unchanged for the last 10 weeks. He had a past history of alcohol abuse, but had abstained for the previous 6 years. Peripheral sensory neuropathy had been diagnosed 5 years previously.

On examination, there was a Parkinsonian tremor (frequency 6 Hz), with typical 'pill-rolling' action at rest in both hands. Tone was normal, there was no bradykinesia and a negative glabellar tap. There was evidence of peripheral neuropathy with reduced cutaneous sensation over both feet, absent vibration sense and absent ankle reflexes in the lower limbs. Examination of the fundi was normal and no other abnormal physical signs were detected.

Initial investigations showed normochromic normocytic anaemia (Hb 10.6 g/dl) and a raised erythrocyte sedimentation rate (123 mm/hr). Thyroid function, serum caeruloplasmin, urinary copper excretion, CAT scan of head and electroencephalogram were all normal. Subsequent investigations showed reduced serum albumin (29 g/l; normal 35-55 g/l) and raised serum globulin (82 g/l; normal 22-33 g/l). The plasma IgM level was raised (76 g/l; normal 0.3-4 g/l), IgG and IgA levels were normal and there were no cryoglobulins. Marrow aspirate showed a dense infiltrate of small mature lymphocytes and plasma cells confirming

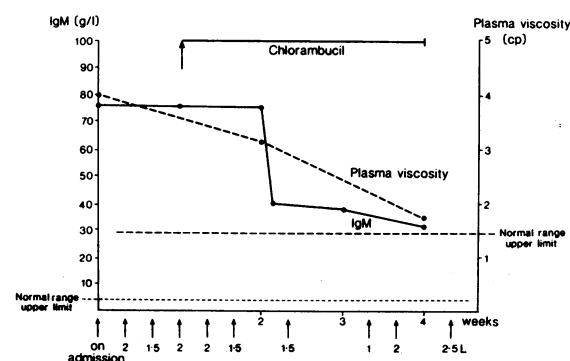


Figure 1. Changes in plasma viscosity and IgM levels with plasmapheresis and chemotherapy over initial 4 weeks of treatment

the diagnosis of Waldenstrom's macroglobulinaemia. Plasma viscosity was markedly raised at 4.006 cp (normal 1.45 cp, 37°C) and treatment was started with plasmapheresis (total of 16 litres exchanged), and chlorambucil 5 mg/day. While immunoglobulin and viscosity levels fell progressively (Figure 1), the tremor remained unchanged, but no further signs of basal ganglia disease developed.

Discussion

The manifestations of Waldenstrom's macroglobulinaemia are due to two pathological processes: (i) IgM is a large, mainly intravascular, molecule which in excess increases red cell aggregation and plasma viscosity. This results in reduced blood flow causing vascular stasis, occlusion and ischaemia and a bleeding tendency due to interference with coagulation factors and platelet function; (ii) plasma and lymphoid cellular infiltrates may result in tumour masses resembling lymphoma or chronic lymphatic leukaemia. The mechanism of the neurological involvement is attributed to various pathologies. Strokes and encephalopathies have improved following plasmapheresis suggesting that reduced cerebral perfusion due to hyperviscosity was responsible⁴. Some of the central effects may be due to haemorrhage or infarction and peripheral effects to ischaemia from red cell agglutination in the vasa nervorum⁵ or deposition of IgM in myelin sheaths⁶.

In the case reported here, the sudden onset and nature of the tremor suggests a small infarct or haemorrhage in the basal ganglia region, although no structural lesion was visualized and there were no other haemorrhagic manifestations. Failure of the tremor to improve despite treatment of hyperviscosity also supports a more permanent lesion. Previously gross tremor of both hands in a patient with Waldenstrom's macroglobulinaemia has been described, but with no obvious Parkinsonian features⁷. The cause of the peripheral neuropathy remains obscure; it is atypical